Research Paper Medical Science

Cholelithiasis in an infant with hypertrophic pyloric stenosis

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ABSTRACT

Objective: To report a case of cholelithiasis with hypertrophic pyloric stenosis in a young infant and review the literature. Methods: A case of hypertrophic pyloric stenosis with cholelithiasis diagnosed during ultrasonographic evaluation is described. Results: A 3 month old infant presented with vomiting from day 7 of life,was evaluated for haemolytic anaemia and diagnosed as hypertrophic pyloric stenosis with cholelithiasis. Predisposing factor for cholelithiasis was starvation and dehydration secondary to pyloric stenosis. The patient was managed with open pyloro myotomy with cholecystectomy. Conclusion: Cholelithiasis in childhood is considered a rare condition, usually associated with hemolytic disease, TPN, starvation etc. The increase in the use of ultrasonography has contributed to an increase in the diagnosis among newborns and young infants. This case report alerts to the possibility of gallstone formation following prolonged starvation.

Keywords :

Introduction

Cholelithiasis in infancy has been reported as a rare condition.(1-4) The occurrence of cholelithiasis in the neonatal period and in young infants has been more frequently described due to the increase in use of abdominal ultrasonography.(4,5) Cholelithiasis in childhood is usually associated with hemolytic disease(.6) In the neonatal period, however, the following predisposing factors have been reported: prematurity; sepsis; total parenteral nutrition; long-term use of furosemide; hemolysis; and congenital anomalies of the biliary tree(.1,4,7,8). Cholelithiasis secondary to haemolytic disease is not uncommon in our population.Howover Neonatal cholelithiasis relatively rare, may be characterized by incidental diagnosis and possible spontaneous resolution(.8-11) Our objective is to report a case of asymptomatic cholelithiasis in a young infant diagnosed incidentally during evaluation for pyloric stenosis. The patient underwent cholecystectomy at the same time of pyloromyotomy.

Case report

A 3 month old female infant a product of consanguineous marriage, born as a full term normal delivery, presented with vomiting of feeds from 7th day of life. The vomiting progressively increased and became projectile. The patient presented late to us at the age of 3 months with features of dehydration and hypochloremic metabolic alkalosis. The electrolyte imbalance normalized over 24 hours with N/2 Saline in D5% and KCl (1:100). The laboratory investigations are tabulated as under:-

Hb	9.4
HCT	31.3
Total Bilirubin	1.08
Congugated Bilirubin	0.2
ALT	70
ALP	159
Total Proteins	5.24
Alb	3.4
Creatinine	0.17
TLC	4950
PLT	159000
Ret	5.79
PBF	Macrocytes, crenated RBCs, Schistocytes seen
pH	7.59
Na^+	129
K^+	3.5
Cl^-	89
Coagulogram	Normal
Blood Culture	Sterile

Haemolytic work up was noncontributory.

Ultrasonography showed multiple stones in gall bladder with pyloric canal length of 15 mm, thickness 12 mm and muscle thickness of 7 mm suggestive of hypertrophic pyloric stenosis. Patient was subjected to exploratory laprotomy through a right subcostal incision. Pyloro myotomy and cholecystectomy was done. Post operative period was uneventful. Normal feeding was resumed in 24 hours. Patient was discharged on 3rd post operative day. Patient is asymptomatic and is on follow up. On analysis the stones were found to contain bile salts (30%), bilirubin (30%), biliverdin (20%) and hematin (20%).

Discussion

Cholelithiasis in infancy is considered rare.(1-4) The youngest child reported by Walker, in a series from The Hospital for Sick Children, Great Ormond Street, of children presenting with symptoms of cholilithiasis, was 4 months (Walker, 1957). During the recent years , incidence of cholelithiasis in infancy has been increasing due to the increase in the use of abdominal ultrasonography in this age group.(4,5,8) Usually, diagnosis of cholelithiasis is incidental, observed in the investigation of other pathologies or in routine examination at neonatal Intensive Care Units.(8-10) Cholelithiasis in childhood is associated with hemolytic disease, congenital anomalies of the biliary tree, infection, and adolescent pregnancy.(4,6,12-15) The following predisposing factors have been reported in cholelithiasis in childhood: prematurity, prolonged fasting, long-term total parenteral nutrition, long-term use of

furosemide, sepsis, dehydration, phototherapy, congenital anomaly of the biliary tree. Down's syndrome, TORCH, family history, and antibiotic therapy are other associated factors. (1,4,5,7,8,10,11,16-18)

Up to 43% of children receiving TPN develop gall stones. The mechanism is probably change of composition of bile and biliary stasis. Because only some patients receiving TPN develop gallstones so other factors also play a role. Sepsis, dehydration, chronic furosemide therapy, cystic fibrosis and short bowel syndrome may also be contributing factors. It is possible that all of the mentioned factors, associated with immaturity of enterohepatic circulation of bile acids, could determine biliary stasis - the main mechanism in formation of gallstones during the neonatal period.(20-22) Other studies have indicated that neonatal cholelithiasis may be a temporary condition due to biliary stasis.(4,11) Prolonged fasting and minimal enteral nutrition may lead to biliary stasis, whereas a full diet stimulates bile flow and, thus, inhibits biliary stasis. Prolonged fasting also inhibits secretion of intestinal hormone, which is responsible for normal enterohepatic circulation of bile acids. The lack of hormone may alter bile composition and, consequently, lead to stasis.(7,11,16) In premature children, total parenteral nutrition is associated with cholestasis and formation of sludge and gallstones.(11) Amino acids play a serious toxic role, since most enzyme pathways of its metabolism are still immature, causing accumulation of intermediary metabolites.(24) The literature underscores the fact that total parenteral nutrition is usually associated with prolonged fasting and minimal enteral nutrition in immature newborn infants. (25,26) Different authors, however, have observed that cholestasis, with consequent lithogenic disorders and formation of gallstones, is a late complication in total parenteral nutrition, especially among premature babies.(27-30) Biliary stasis favors proliferation of bacteria, and thus causes an increase in the production of beta-glucuronidase enzyme, which is a product of bacteria. This enzyme is responsible for hydrolysis of conjugated bilirubin into unconjugated bilirubin. A greater amount of unconjugated bilirubin increases saturation of the bile, causing formation of gallstones. (8,17,21,31) The use of diuretics, especially of furosemide, inhibits transportation of sodium into the cell interior, which may cause a reduction in excretion of bile acids and, consequently, the formation of gallstones. According to the literature, incidentally diagnosed asymptomatic neonatal cholelithiasis usually has a benign course, with about a 50% chance of spontaneous resolution of cases during the first 6 months of life(.7) Resolution of cholelithiasis occurs after the dissolving of gallstone or after fragmentation and passing of gallstone through the biliary tree.(35) Long-term follow-up is recommended for children persisting with gallstones after 6 months of age, as long as they have remained asymptomatic.(7,35 Surgical intervention should be reserved for symptomatic cases or those in which calcification of gallstone occurs(.36,37) The case presented in this report is in agreement with the literature with respect to presence of the following risk factors: prematurity, sepsis, total parenteral nutrition, longterm use of furosemide, delayed introduction of enteral nutrition, use of phototherapy, and mother with history of preeclampsia. The diagnosis of our patient was incidental; it occurred during a routine follow-up,evaluation and ultrasonography examination of infantile hypertrophic pyloric stenosis. In the study by Randall et al.,(34) formation of gallstones in high risk-group patients occured primarily during the first 12 weeks of life. In our case, the patient was asymptomatic, but incidental cholecystectomy was performed simultaneously while performing Ramstedt's pyloromyotomy. Some authors have only recommended cholecystotomy on children suffering from cholelithiasis, provided there are no inflammatory changes in the wall of the gall-bladder (Chiray, Debray and Delattre, 1950; Gross, 1953: Grob, 1957). Experience with adults shows, however, that the gall-stone disease in these cases often recurs and requires renewed operation. This has been reported also from observations on children who have been subjected to cholecystotomy (Edington, 1930; Barnes, 1958).

In our opinion, gall-stones disease in children does not in any essential way differ from that in higher age groups. Soderlund and Zetterstorm had similar observations from their study. No intraoperative or post-operative complication occurred in our patient.

PICTURE OF INDEX CASE

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